CASE REPORT
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MUCORMYCOSIS – "A NUISANCE IN THE IMMUNOCOMPROMISED PATIENT"

Maxillofacial Surgery

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ABSTRACT

Background: The aim of this case report is to present an interesting case of fungal infection secondary to osteomyelitis of maxilla. **Methods:** A female patient, 42 years old, reported to the Department of Oral & Maxillofacial Surgery, I.T.S Centre for Dental Studies & Research, Murad Nagar, India, complaining about pain & bad breadth from her oral cavity. Her medical history revealed she was under antidiabetic medications from previous 2 years. Subsequently, the cone beam computed tomography examination revealed large mixed-density lesion in alveolar process of maxilla, giving the impression of moth-eaten appearance. The clinical examination revealed exposed necrotic alveolar bone in upper left maxillary premolar region. **Results:** Based on the clinical & radiological examination, a treatment plan for surgical intervention which includes extraction of involved teeth with sequestrectomy followed by infrastructural maxillectomy was made & was implemented. And after histopathological diagnosis of chronic osteomyelitis with fungal infection, antifungal regimen was started with Amphotericin B.**Conclusions:** Individuals having compromised immune systems have more chances to get lethal fungal infections that affect the oral cavity. So, any osteomyelitis in maxilla must get diagnosed at early stages to prevent the terrible outcomes.

KEYWORDS

Osteomyelitis, Infection of jaw bones, Sequestrum, Maxillectomy, Mucormycosis

INTRODUCTION

Osteomyelitis is defined as the presence of exposed bone in the oral cavity, which is failed to heal even after providing adequate intervention.[1] Contemporary literature on infections of the face and skull bones concur that the maxilla is the second most prevalent place to develop osteomyelitis, beneath the frontal bone. This can be attributed to its prominent site and peculiar morphology, which places spongy bone in close vicinity to other tissues that may harbor a primary infection, including as the teeth, gums, and the antrum of Highmore.[2] Osteomyelitis, or bone inflammation, develops as a medullary cavity infection, quickly involving the haversian systems and extending to the periosteum.[3] In immunocompromised people, osteomyelitis frequently develops as an adverse manifestation of odontogenic infections.[4] It generally impacts the long bones and vertebrae; rarely, the cranial bones are involved.[5] Since the maxilla has a wide collateral blood flow, thin cortical bones, and strutted bone marrow that reduces infection risk, it tends to happen there more frequently in the mandible.[3] In 1852, Edouard Chassaignac, a French physician, presented the first description of osteomyelitis. Rees (1847) described maxillary osteomyelitis.[6] Factors which contribute to osteomyelitis are systemic diseases which compromise the immune system of an individual such as diabetes mellitus, HIV, malnutrition, and use of chemotherapeutic agents.[3] According to Macbeth (1952), the etiology of maxillary osteomyelitis can be broadly divided into three categories: traumatic, rhinogenic, and odontogenic.[6] Here, we report an uncommon case of maxillary osteomyelitis in an adult lady who had a history of diabetes from previous 2 years, with rare atypical clinical presentation and was diagnosed, treated, and regularly followed up.

Case Report

A 42-year-old lady came to our oral and maxillofacial surgery outpatient clinic complaining of pain and bad breath emanating from her mouth that had persisted for ten months. Pain was lancinating localized, aggravating while taking food, and she took medication (analgesic) from the local medical store, and the pain subsides. She also noticed a pus discharge, clusters of exposed yellow particles that looked like structures, and an unpleasant smell. History of diabetes from previous 2 years was also given by the patient. Genealogical background was unrelated. The patient had no deleterious habits, according to her personal history. Regarding the extraoral and general physical examinations, a diffuse swelling having color, homologous to adjacent skin and of size measuring around (2×2) mm extending below the infraorbital rim at the superior extent till the ala of nose having inferior extent and antero- posteriorly extending from nasolabial fold to pre auricular region. No positive history of paresthesia noted over that region. Exposed necrotic alveolar bone was detected during an intraoral examination, and it was extended from the upper buccal vestibule involving the left maxillary premolar region till maxillary 1st molar mesiodistally (Figure 1).

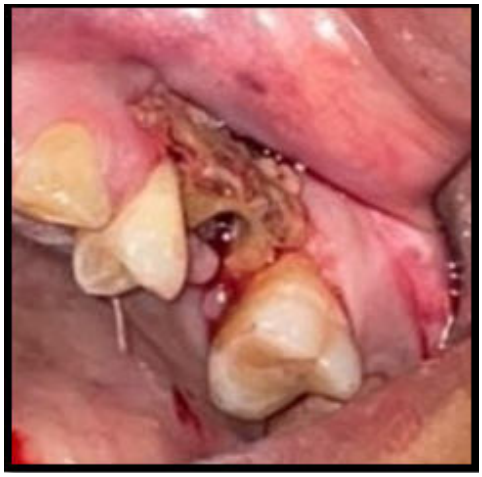

Fig.1 Exposed Necrotic alveolus in maxilla

Also, there was grade 3 mobility of 1st premolar on same side was appreciated. A tentative diagnosis of chronic suppurative osteomyelitis of the maxilla was made considering the clinical symptoms. For radiological examination, a Cone beam computed tomography (CBCT) scan was conducted. In axial view, results showed, a large mixed-density lesion covering the maxilla's whole alveolar process, giving the impression of moth-eaten appearance. Also, on the left side, a maxillary sinus breach was seen, suggestive of mucosal thickening in the coronal view (Figure 2).

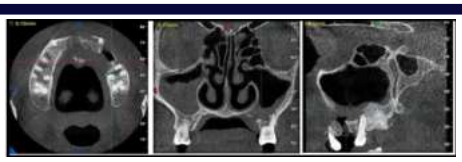

Fig.2 Axial view showing mixed density lesion with moth-eaten appearance, Coronal view showing breach in maxillary sinus & Sagittal view showing thickening of maxillary sinus

The aforementioned characteristics all pointed towards left-side maxillary osteomyelitis. A treatment plan of surgical intervention which includes extraction of involved teeth with sequestrectomy followed by infrastructural maxillectomy was made. All other necessary blood investigations and random blood sugar test was done prior to surgery. Patient was admitted in the ward of oral & maxillofacial surgery of I.T.S Dental College, Murad Nagar. Consent for surgical intervention was taken from the patient prior to surgery. A prophylactic coverage of 1.2gm Amoxicillin (1000mg) with potassium clavulanate (200mg) was given 1 hour before the procedure. Painting & draping was done under aseptic condition. Under local anesthesia, extraction of all the involved teeth & sequestrectomy was performed followed by infrastructural maxillectomy. Defect was debrided with betadine and saline & an iodoform dressing was given. After which patient was on regular follow ups & iodoform dressing was changed after every eighth day. An obturator was made to close the surgical defect (Figure 3).

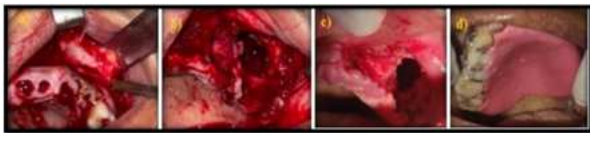

Fig.3 (a) Extraction of involved teeth, (b) Sequestrectomy followed by infrastructural maxillectomy, (c) Reduced defect size on follow up periods, (d) Obturator covering the defect

And specimen was sent for the histopathological examination (Figure 4).

Fig.4 Specimen sent for histopathological examination

The patient was kept on nasogastric tube feeds for 7 days and thereafter started oral feeds. Therapeutic antibiotic course of 1.2 gm Amoxicillin (1000mg) with potassium clavulanate (200mg) & Metroniadazole (500mg) and Diclofenac (75mg) as an analgesic was given through systemic route for next 5 days.

In histopathological report they mentioned that the submitted H&E-stained section shows loose fibro cellular connective tissue stroma showing irregular bony trabeculae with empty lacunae and no evidence of peripheral osteoblastic rimming suggestive of non-vital bone. The connective tissue stroma shows thick and thin collagen fiber bundles interspersed with varying calibers of blood vessels. The stroma also shows the presence of non-parallel, non-septate broad hyphae with empty bulbous swollen areas within them. The non-septate hyphae show focal branching at acute to right angulation. Focal areas show necrosis surrounded by inflammatory infiltrate chiefly containing neutrophils. At focal areas hyphae are appreciated in close proximity to the blood vessels and bony trabeculae. Fungal spores are seen within the hyphae and lying freely within the connective tissue stroma. The hyphae were positive for PAS and GMS stain. A final diagnosis of "chronic osteomyelitis of the maxillary bone with fungal infection (mucormycosis), causing extensive tissue necrosis" was made based on these histological characteristics.

We further obtained a physician referral for management of blood glucose levels and for systemic antifungal therapy with Amphotericin B.

DISCUSSION

One of the most difficult infectious disorders to cure is osteomyelitis, but there are certain additional characteristics that make it more difficult to treat. Comorbidities such as diabetes, radiation, starvation, anemia, cancer, hypertension, and immunosuppression all have an impact on immune surveillance and impede the healing of wounds. Osteomyelitis and diabetes mellitus are closely correlated conditions that are known to inhibit the host immune response. Reduced leukocyte chemotaxis and phagocytosis with reduced vascularity, which lowers tissue perfusion, is one of the mechanisms that promotes bone infection thereby aggravating osteomyelitic process. In addition to lowered immunity and altered vascularity, ketone bodies in diabetes people promote the development of fungus. The fungus produces the enzyme keto- reductase, that acts by interacting with ketone bodies. [7] Rhizopus arrhizus causes mucormycosis in a high percentage of diabetic individuals because it produces the enzyme ketoreductase, which enables the organism to use the patient's ketone bodies.[8] Changes in pH and capillary permeability following bacterial inoculation led to various consequences such as tissue degradation, leukocyte recruitment, reduced oxygen tension, increased local pressure, small-vessel thrombosis, and bone degeneration. Increased pressure as the infection develops into the medullary cavity causes Haversian and Volkmann canals to spread it into the cortex. This spread then causes periosteal stripping, which results in the loss of the periosteal blood supply and ultimately causes bone resorption and necrosis.[3] No matter what the concentration level is, it is typically difficult to reach the target area because the delivery of antibiotics in the dead bone is so sluggish.[6] Options for treatment range from several straightforward non-invasive techniques to more extensive and invasive procedures. Antibiotics, NSAIDS, hyperbaric oxygen therapy, bisphosphonates, and muscle relaxants are all part of the nonsurgical approach. When a nonsurgical method fails, surgical options to take into account are segmental resection, partial (marginal) resection, decortication alone, and decortication combined with bone grafting. A medical professional finds himself with a dilemma because an aggressive care may result in significant co-morbidity and the consequent need for reconstructive surgery, while conservative management almost always leaves an opportunity for repeated recurrences of the illness.[9]

CONCLUSION

According to literature, osteomyelitis is the most common and oldest disease. Individuals with impaired immune systems are susceptible to lethal fungal infections that affect the oral cavity's soft tissues and bones. Accurate diagnosis should be made with consideration through examinations like radiographic and histological analysis. For diabetics, maintaining optimal glycemic control is essential to preventing these infections. An individual may experience major complications from a maxillary infection, including brain and cranial cavity infections. Any maxillary osteomyelitis must get immediate, aggressive treatment to prevent the terrible outcome.

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Conflicts Of Interest

No conflict of interest.

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